

# Ethics, justification and the prevention of spina bifida

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During the 1970s, prenatal screening technologies were in their infancy, but were being swiftly harnessed to uncover and prevent spina bifida. The historical rise of this screening process and prevention programme is analysed in this paper, and the role of ethical debates in key studies, editorials and letters reported in the *Lancet*, and other related texts and governmental documents between 1972 and 1983, is considered. The silence that surrounded rigorous ethical debate served to highlight where discussion lay—namely, within the justifications offered for the prevention of spina bifida, and the efficacy and benefits of screening. In other words, the ethical justification for screening and prevention of spina bifida, when the authors are not explicitly interested in ethics, is considered. These justifications held certain notions of disability as costly to society, with an imperative to screen and prevent spina bifida for the good of the society.

ethics in the hope of hearing something new, something that also tells a story of medical ethics. This paper explores medical ethics in action through the analysis of papers from the *Lancet* between 1972 and 1983 related to the development of prenatal screening for spina bifida.

We focus on a select group of sources and on a particular medical problem. The *Lancet* was chosen as it represents a particular medical perspective in a period in which prenatal screening was burgeoning, and because it was (and is) regarded as an authoritative and widely read journal. Because of its status within the medical community, it also offered an insight into key studies and discussions, allowing us to outline changes in medical knowledge surrounding prenatal screening. As Treichler has shown, a field can be quickly constructed, strengthened and controlled by the work of a few key medics and the status of spina bifida within the medical community also demonstrates this insight.<sup>3</sup>

From the outset, we want to qualify our findings. It would be foolish to claim that the *Lancet* authors represented all the opinions expressed in relation to prenatal screening for spina bifida. We are limited by the fact that these papers may not express the author's full opinion in this matter, and what is written could reflect a particular genre of medical writing that hindered expression. However, studies often did offer justifications for screening and personal opinions were articulated. In addition, letters were published in response to studies, and editorials reflected the views of the writer. Although there is no doubt that editorial convention helps to shape the writing of any author, what they chose to say came to affect the way screening was performed.

We expected that the primary focus of what was named as ethical considerations would be issues around the doctor–patient relationship; yet, the justification of prenatal screening for spina bifida was not based in the principle of this relationship. Instead, these papers offer us a chance to consider how ethics was expressed historically through the justification to screen for spina bifida. In any medical study, there is always a validation for the study by an appeal, either implicit or explicit, to its rational and social value. We aim to understand what these justifications and guiding values were during the emergence of new screening modalities. We find that justification for prenatal screening was implicit in the *Lancet* between 1972 and 1983, and were grounded in terms such as prevention,

In 1975, it was reported in the *Lancet* that “the finding that AFP [alpha-fetoprotein] levels are often raised in maternal blood in association with neural tube defect of the fetus is an important advance in obstetric practice since it presents the possibility of a screening programme leading to early diagnosis and termination of these abnormal pregnancies.”<sup>1</sup>

In Britain, during the 1970s, prenatal screening underwent a revolution in the form of ultrasonography and alpha-fetoprotein (AFP) screening. As this quotation suggests,<sup>1</sup> these new technologies were seen as a significant step forward in identifying abnormal fetuses and as a tool to facilitate the possible termination of pregnancy.

In this paper, we offer a historical perspective on medical ethics, but one that differs from the majority of work devoted to the history of medical ethics. For the most part, the history of medical ethics has focused on the development of the discipline itself. Thinkers such as Albert Jonsen<sup>2</sup> specifically consider the key events, situations and legal frameworks within which the discipline developed, or the key factors influencing the thinking of bioethicists. In this sense, the history of medical ethics and the emergence of bioethics is a history of high medical ethics. In other words, these histories deal with what could be named as medical ethics in itself.

Although this kind of work is no doubt important, we offer a different notion of the history of medical ethics by examining the justifications for the screening of spina bifida. Here, we propose to listen to the silences around

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**Abbreviations:** AFP,  $\alpha$ -fetoprotein; DHSS, Department of Health and Social Security; NHS, National Health Service

efficacy and benefit. Through a close examination of these charged terms, the justification of mass programmes to screen for spina bifida emerges as one that embraces a complex economic morality.

## NEW TECHNOLOGIES AND THE NOTION OF PREVENTION

Two main methods of detection were used to screen for neural-tube defects—ultrasonography and AFP testing. Ultrasonography was already in military use in the early 20th century, and was used as a diagnostic medical tool in America in the 1940s. Innovations occurred in Glasgow under Ian Donald during the 1950s, but it was not until the 1960s that it was applied more regularly when studying the fetus. By the late 1960s, ultrasonography was first used to uncover fetal abnormalities, and Stuart Campbell seems to be the first to have diagnosed an anencephalic fetus at 17 weeks in 1972 and spina bifida in 1975.

In tandem with these sonographic developments in 1972, Brock and Sutcliffe realised that the increased AFP levels in amniotic fluid were associated with fetal abnormality. By 1974, maternal serum was being used clinically to detect AFP levels as a predictor of fetal abnormality, and by 1977, a screening regimen was in place for women who were at 16 and 20 weeks of gestation (optimum time between 16 and 18 weeks). The screening programme began with maternal testing of serum for AFP levels, followed by ultrasound and amniocentesis for screening of amniotic fluid, with a termination that could be speedily arranged. Those deemed at risk of giving birth to a child with spina bifida underwent amniocentesis without serum screening.

Childbirth was already a legitimate moment of medical intervention, but this was also a period in which pregnancy was in the headlines because of the Abortion Act of 1967, the rise of family planning and the contraceptive pill. Medical involvement also made sense here as fertility rates fluctuated and overall child mortality fell dramatically. But something new was afoot. With the rise of screening for a spina bifida baby, a space opened for a shift in focus from preventing death to uncovering abnormality.<sup>4</sup>

## THE ETHOS OF PREVENTION

The rise of preventive medicine in relation to public health has been well documented by many historians. Although we do not examine this well-worn narrative, it is important to note that from the rise of preventive medicine from the ashes of the 19th-century sanitary ideal, technology and medical innovation were clearly important points of rational quantification and evaluation that increased state intervention in everyday life.<sup>5</sup>

The creation of the National Health Service (NHS) in 1948 saw public health as both a national responsibility and a priority.<sup>5</sup> By the 1970s, public health was increasingly on the government's agenda. The NHS's reorganisation in 1974 helped to emphasise the fight against ill health by promoting personal responsibility.<sup>4</sup> In the struggle to rationalise this approach, the concept of prevention became a powerful tool. Used in a series of governmental documents—all beginning with the title "Prevention"—smoking, drinking, eating habits, and pregnancy and childcare were of central concern. Embedded in these texts was a desire to reorient behaviour to, "keep people healthy and to improve the quality of life".<sup>6</sup> Such a vague statement warrants further consideration as it fails to examine what governs the choices made in preventive medicine, whether in terms of categorisation or treatment. Yes, screening sprang from the medicalisation of the motherhood, the rise of technology and conceptions of disability, but also from the fact that categorisation of illness outstripped ability to cure.<sup>7</sup> Spina

bifida was detectable but not curable. Skrabanek<sup>8</sup> has defined this form of prevention as "anticipatory medicine", which speculates the possibility of risk. In terms of prenatal screening, this is a complex issue, as it is difficult to tell whose health takes centre stage: the mother's, the child's or the nation's. With screening, prevention would take on a new meaning.

The health of the nation and prevention were entwined in governmental documents. When uncovering abnormality, they suggested that "the only 'treatment' on offer is termination of pregnancy".<sup>9</sup> This ethos was found in the *Lancet* texts. Of course, work had been carried out to discover the roots of spina bifida; race, environment and even potatoes had been put forward as likely, if unsound, candidates, but termination as prevention was even being promoted by Brock and Sutcliffe, the pioneers of AFP screening in 1972. In a remarkable short piece that covered just two pages in the *Lancet*, they stated thrice that their screening tool would allow for the termination of those with anencephaly and spina bifida:

A marker molecule, which indicates an affected fetus early enough to allow termination of pregnancy, has so far not been found. We suggest that alpha-fetoprotein (AFP) could act as such a marker molecule.<sup>10</sup>

What is interesting about this quote is the accepted coupling of "affected" pregnancies and "termination". Brock and Sutcliffe's primary goal was to find a marker and an optimum moment that enabled a distinction to be made between abnormal and normal fetuses in order to facilitate the termination of the abnormal. To many, this may have seemed an obvious link, but by unquestioningly suggesting that abnormality led to termination, medicine over-reached its boundaries, perhaps crossing the brink into social engineering. Although this is no simplistic accusation of eugenic principles at play, for some, this idea was so embedded in medical practice and notions of progress that no discussion of therapeutic options or choices was made.

This position was apparent in a number of influential works in the *Lancet* such as that of Campbell *et al.*<sup>11</sup> Campbell thought that ultrasound could enhance AFP screening. In Campbell's study, after raised AFP levels were recorded, ultrasound was used as a tool to confirm the presence of abnormalities. In Campbell's series of three case studies, all ended in a termination recommended by the medical staff. The efficacy and benefits of this procedure is discussed later, but it is interesting to note that the link between detection of abnormality and termination was so strong that termination was recommended in all three cases rather than offered as one alternative, even though one fetus was noted to have no sonographic evidence of spina bifida or anencephaly.<sup>11</sup> Although reaction to the efficacy of ultrasonography was evident, little was said in relation to the correlation between abnormality and termination.

Yet the natural link between abnormality and termination was not only to be found in Brock and Sutcliffe, or in Campbell. Most saw this as a clear indicator of scientific advancement; their excitement and even pride could not be contained. Leek *et al.*<sup>12</sup> proclaimed, "this is the first reported case of prospective diagnosis and termination of an open neural tube defect arising from routine screening".

These views were consolidated in the influential report of the UK Collaborative Study on AFP in relation to neural tube defects in 1977.<sup>13</sup> Again, termination was offered as "the only means available for reducing the number of live infants born with these congenital defects". As confidence grew in the process, and serum testing became an option, widespread screening became a distinct possibility. This was not an

outlandish prospect, as screening in “high-risk” cases was already routine even though no one could clearly delineate who was really at risk, let alone at high risk.<sup>14</sup> Still, the Department of Health and Social Security (DHSS) strongly endorsed this agenda.<sup>13 15</sup>

## CONVERSATION AND THE EMERGENCE OF THE ETHICAL

A few voices did ring out against the coupling of termination and prevention in letters to the editor of the *Lancet*. One such letter, by Brereton, highlighted several pertinent points as follows:

- (1) Termination did not prevent abnormalities, but prevented the birth of an abnormal fetus, a claim also made by Goodhart.<sup>16</sup> This did not mean that termination was seen as objectionable merely because primary prevention could prevail at some point.<sup>17</sup>
- (2) There was a lack of delineation in relation to abnormalities. This was patently clear in most studies in which “abnormality” or “malformation” were used as catchall terms. Another letter by Ellison-Nash<sup>18</sup> really argued this point effectively.
- (3) The decision-making process could be problematic.<sup>19</sup>

Although none of the above points received rigorous ethical debate, Brereton’s concern over decision-making processes did begin to explore, however weakly, the ethics.

Other *Lancet* authors, such as Walker, tried to emphasise that decisions to terminate pregnancy should be made jointly between the parents and the obstetrician. This was merely a restatement of the importance of decision making, which was the dominant discourse in medical ethics at that time.<sup>20</sup> Likewise, other than a single editorial remark and a few observations about “distaste” for termination in some hospitals, and religious or moral obligations in stranded and perplexing sentences, the question of decision making remained unworthy of lengthy discussion in the *Lancet*.<sup>14 17</sup>

However, the question was raised in one governmental document, the DHSS consultation paper, “Screening for spina bifida and other neural tube defects”, which was referred to by one article in the *Lancet*.<sup>15</sup> Still, in the section devoted to “Ethical Problems”, this discussion was very brief—five sentences in length. The ethical qualms were represented here in terms of the difficulties that arose for the doctor–patient relationship, only insofar as the doctors’ decision might be challenged by the patient, as stated, “difficulties may arise when there is reason to suppose that termination of pregnancy would be unacceptable”.<sup>21</sup> Issues over the doctor–patient relationship was firmly placed back into the hands of the practitioner.<sup>21</sup>

Consistent with what we expected to find, the only moment named as the ethical was when a doctor’s decision might be questioned. In other words, the ethical is only named as such when the desired outcome—termination—was questioned. Such doctor-centred decision making is not surprising during a period in which medical paternalism was still commonly acceptable. It would be anachronistic to dwell on the lack of discussion about the importance of patient-centred decision making; instead, we wish to draw attention to the other two points. Firstly, the drive to prevent the birth of abnormal fetuses through screening and the subsequent termination was deemed as progress—scientific, medical and social development. This was indicated by the coupling of diagnosis and prevention, abnormality and termination. Secondly, the risk was only addressed in relation to normal fetuses that might be lost during amniocentesis and the problems

surrounding efficacious screening, which we deal with in the next section.

These short conversations within the DHSS document addressing “ethical problems” serve only to highlight the paucity of ethical discussion within the *Lancet* itself. Although we may not expect the *Lancet* to have sustained ethical discussion, what is most interesting to us is the fact that ethics does emerge, but only in conversational tones, and that screening only became an ethical question when it was perceived to hinder scientific and public health progress.

Cooter<sup>22</sup> has suggested that in “most exercises in the application of philosophical logic to “practical” medico-social issues, one comes away dismayed at the shallowness (or absence) of socioeconomic and political understanding, at the technological determinism behind the ethical agenda setting ..., and at the underlying uncomplicated notions of, and faith in, “progress” and change”. This statement is harsh indeed, but it is possible to consider this point in another way—namely, that the lack of a rigorous notion of an ethical dimension results from more robust and unquestioned notions of socioeconomic and political progress. It seems more likely that ethics was conceived as unquantifiable, such as, the doctor–patient relationship, which did not lend themselves to numerical designation and quantifiable markers of progress. Implicit in the *Lancet* and related papers is a sense that the authors thought they were making calculations independent of ethics or morality.

In addition, the same DHSS document draws our attention to the distinction between the quantifiable and the non-quantifiable, which it couches as the humanitarian versus the economic. In some senses, it seems that they are binary opposites as the opening paragraph of this section reads:

While the humanitarian arguments for the prevention of spina bifida and related disorders are paramount, the economic considerations also deserve examination.<sup>21</sup>

The rest of the four-sentence paragraph deals solely with economic concerns of universal screening and the prevention of costs incurred in the care of children born with spina bifida. We find no evidence that these “humanitarian arguments” for screening were being offered.

However, unlike prenatal screening, such debate was evident in the “treatment” of spina bifida, specifically in the case of “selective” treatment. A discussion between Lorber<sup>23</sup> and Zachary<sup>24</sup> centred upon the justification of treating such infants. Zachary’s promotion of treatment for all came up against Lorber’s belief in selective treatment for those deemed a burden. Here, the question of personal, familial and social suffering was expanded upon in detail. In the early 1980s, this debate was also explored by Harris, Anscombe and Cuisine, who engaged with Lorber on the ethics of what Harris termed “selective non-treatment” that effectively advanced the death of infants with spina bifida considered too disabled to live.<sup>25–28</sup> What is interesting is that the *treatment* debate did not overtly leak into discussion over screening and prevention. This lack of dialogue within prevention suggests perceived differences between prevention and treatment, and a fetus and a live baby.

In addition, there was activity around disability rights in the 1960s and 1970s. Still, there is scant evidence that early screening debates heard these voices. Davis,<sup>29</sup> the activist, did attempt to engage with the medical arena during the 1980s, but seems to have had little impact on early screening debates. In reality, the justification for the prevention of spina bifida births came down to a particular balancing of efficacy and benefits, and it is here that the justification for screening was forged.



Our attention then is drawn not to what is named as ethics in itself, but instead to the silences about ethics. Ethics, at least in part, is about the justification of a decision to act in a certain way. Scientific and technological progress, as noted above, is its own assumed justification, but is also tied up with notions of societal progress. We turn then to the bulk of the discussions in the *Lancet*, which were mostly based on issues surrounding efficacy, which was constructed with preconceived notions of benefit in mind. What was deemed as an exercise in ensuring efficacy and cost:benefit analysis—the economic as opposed to humanitarian/ethics—was actually a process of moral justification, as we shall now show.

## UNCOVERING THE ETHICAL Efficacy

In contrast with the discussion labelled as ethics, debate raged around the question of efficacy. Authors spent considerable energy weighing up whether the process actually detected the abnormal, and, secondly, whether this was an efficient way to carry out the task of screening. Several key topics were considered including the line at which AFP levels denoted abnormality, when affected fetuses were missed, the impact of screening on normal births, factors that affected readings, who to screen and finally issues that lay outside of scientific control. These were seen as worthy of discussion as they were points of clear dispute, but were also expressions of preformed notions of disablement.

Brock and Sutcliffe's AFP measurement signified a decision that amounted to the drawing of a line. As was stated in the 1977 collaborative report, there was no natural level of AFP but instead there were measurements that required expert reading.<sup>13</sup> On one side of the line were those with supposedly normal AFP ranges, and on the other were those with high AFP levels; one side normal and the other abnormal. Discussion rested on the most efficacious point that such a line could be drawn rather than on questioning the validity of such an artificial demarcation of disability.

After much discussion, it was agreed that AFP levels that exceeded the "normal median" by 2.5 times at 16–18 weeks of gestation was defined as abnormal.<sup>13–15</sup> Apprehension existed as to the number of impaired that would be missed if the line shifted to the right, but real anxiety was allocated for those healthy infants who would be terminated, as there are always false positives. A repeated screening may have weeded out incorrect readings, but it was possible for high AFP ranges to be consistent with unaffected births.<sup>30</sup> For example, in the study by Campbell *et al.*,<sup>11</sup> a normal baby was terminated because of raised AFP levels in both the maternal serum and amniotic fluid, and because the mother had previously produced an affected baby, despite the lack of ultrasonographic evidence of spina bifida. Leighton *et al.*<sup>1</sup> suggested that 5% of cases would prove to be false positives, although this was decided via statistical analyses rather than material data. The regional incidence of spina bifida should have been an important factor, as Wales and Northern Ireland both had high rates of "abnormal" babies. Conversely, areas with a lower incidence of affected fetuses could artificially raise the numbers of false positives, and it was suggested that screening could harm more healthy fetuses than abnormal ones. This issue was never fully discussed in terms of screening programmes, but the desire to target those who were thought to be at high risk was discussed.

Usually the most efficacious way to find affected fetuses would be to screen high-risk groups. The concept of risk was fundamental to the New Public Health. Expertly defining statistical risks was seen as a neutral procedure. In terms of the estimation of risk—or should it be called the construction of risk categories—in the New Public Health, Castel<sup>31</sup> has shown

that the new definition of risk gave rise to moments of legitimised intervention while being sold as unproblematic. In a period where cost was increasingly important, it was vital to target those seen as posing the greatest economic risk.

From the beginning, those who were deemed most likely to produce an abnormal baby underwent an amniocentesis without serum testing. At first glance, this would seem a logical procedure until it was realised that only 10% of affected births were associated with those who were categorised to be at high risk. A total of 90% of affected births were from those with no previous history, or from other risk categories. Thus, the targeting of those deemed at risk was problematic and was not a particularly successful way of preventing such births. Moreover, assumptions could be made about the likelihood of having a baby with spina bifida that could lead to false readings, as the Campbell case indicates.

Incidents of spina bifida had been higher in Britain than in some other countries, which did help to spur on governmental concern. Here statistics could lead the way. In 1975, the DHSS recorded 1748 children with a neural-tube defect. Looking at spina bifida alone, and by taking into consideration a range of viewpoints, this was translated into 2.6 births/1000 in 1975. Moreover, the incidence of spina bifida was decreasing before the routine screening of all women was in place, although the screening of high-risk pregnancies may have helped shape these figures. Falling per year from 1138 in 1970 to 979 in 1973 the incidence had decreased to 678 by 1976.<sup>32</sup> The reasons behind these drops were unclear and, some believed, did not reflect an increase in screening.<sup>32</sup> In reality, such figures were problematic, and did not take into consideration regionalism and fluctuations in incidence. For Althouse,<sup>32</sup> the only way to make sense of screening was to look closer at the efficacy of screening and ignore national figures. Althouse, however, seemed to ignore the fact that efficacy figures only served to highlight effectiveness (not whether this procedure was needed in the first place). The need was assumed. Strangely, no one seemed too interested in the innovations in treatment for spina bifida during this period, in the form of valves to control cerebrospinal fluid pressure and operations to close lesions. Again, these developments were found within the selective treatment debate and did not form part of an ethical discussion within screening.

Although problems surrounded the idea of a line to distinguish abnormality, high-risk patients were no real benchmark of impairment, false positives could occur and the level of defect was unknown, screening was also subject to the vagaries of practice. Other factors, such as twinning, blood in the amniotic fluid sample and screening at the wrong time could all artificially increase serum and amniotic AFP levels. To some extent these could be uncovered by ultrasound, but that was not in the least certain as Campbell *et al.*'s study showed.

In addition, interpretation of AFP levels was dependent on the accuracy of menstruation dates, putting the onus on women for the efficacy of a screening and termination programme. Moreover, Chamberlain<sup>33</sup> noted that women did not always undergo screening at the optimum time; they refused a termination or they declined further tests. Oddly, the reluctance of parents to go through the full process was seen as a hindrance to efficacy of the prevention programme, rather than a point at which genuine ethical discussion could take place.

Moreover, parental concerns were interpreted as affecting the success of prevention of spina bifida, materially and negatively. In Roberts *et al.*'s<sup>15</sup> study in South Wales in 1983, Chamberlain's concerns were substantiated. The failure of women to undergo screening or agree to a termination caused a decrease in efficiency levels in practice from 95% down to 65%. No one

discussed why women would choose not to undergo screening and termination except in simplistic and conversational tones.<sup>15</sup> Roberts' discussion focused on "failure to undergo screening", where open neural-tube defects were being missed and gestation dates were problematic. Although he considered the role of the practitioner and improvement in administration as being important factors, the failure to terminate was described as "another disappointing finding ... arising from an aversion to abortion and fear of termination of a normal pregnancy, the high false positive rate of serum AFP tests, and the fact that some 60% of OSB [open spina bifida] pregnancies end in stillbirth or neonatal death".<sup>15</sup>

In short, Chamberlain and Roberts *et al* saw that patients avoided invasive medical procedures, but failed to see it as an issue worthy of discussion beyond the need for better education and communication. No one seriously examined the possibility of any moral concerns that patients might have had. In reality, it was never clear exactly how many fetuses would really be detected. Chamberlain and Roberts had shown that many factors interfered with the screening process and no amount of cost analysis would account for human decision-making processes. This was perceived to be a problem in a period in which public health had promoted personal responsibility. Although the incidence of spina bifida was not due to personal choice, screening was. Mothers could be blamed if they failed to adhere to the recommendations of the medical profession, failed to turn up on time, mistook their gestation dates and, even worse, if they refused to undergo a termination when positively diagnosed.

In terms of efficacy then, there was much argument about the process of screening. Issues focused on the reliability of this process and the confounding variables that would impinge on the numbers of fetuses with spina bifida that could be found and terminated. It becomes clear that numbers of false positives and the ambiguity of an abnormality line were not enough to halt screening programmes. The desire to uncover the disabled was enough of a benefit to support technology being harnessed in this manner, independent of the vagaries of efficacy. As MacIntyre<sup>34</sup> points out in *After Virtue*, which was published shortly after these studies, the great moral imperatives of modern society are the efficacy and efficiency of a process. The end, the goal or the presumed benefit is never questioned. Progress in science and society is its own justification. The debate around efficacy expressed certain preconceived notions of benefit, as we shall see this was articulated as the prevention of what was understood to be a costly and non-productive future member of society.

## Benefit

It is clear that the biggest benefit of prenatal screening was the prevention of the births of those with spina bifida, but why was spina bifida seen as so problematic? In these studies, the problem of spina bifida was assumed and a simple correlation between prevention and benefit was assumed. However, some tried to quantify the possible benefits via cost. Here, cost was seen in light of both utilitarian principles of happiness and also in terms of economic/monetary value. Of course, these two issues overlap, but for the purposes of this paper, and indeed many authors attempted to do just this, they are divided into two.

Financial cost is at the core of understanding the position of disability in society from the late 18th century until today. Here the rise of industrialisation has been coupled with the understanding of the economic accountability of an individual. In terms of disability, writers such as Michael Oliver<sup>35</sup> have suggested that impaired bodies were seen as a drain on the economy rather than a source of production. This characterisation helped to medicalise the impaired body. In reality, the

construction of disability was more complex than suggested here, but the rise of industrialisation and the role of the medical profession were significant and interlinking factors.

Cost was an important issue during the emergence of AFP screening, and was acknowledged in the *Lancet*, as well as in the Department of Health documents. A piece entitled, "How to set priorities in medicine" stated that, "the allocation of priorities in medicine—for money, manpower, and materials—is inescapably the most important topic facing the profession at this time".<sup>36</sup> This was more extensively discussed by Meade,<sup>37</sup> who saw that cost and balance were now fundamental parts of any medical discussion because of three problems: (1) the rise of knowledge and technology to support life; (2) NHS consumers had risen in number; and (3) that the economy had worsened. Indeed, the rise of screening and prevention agendas occurred during a period of economic difficulties, in which rates of inflation and oil prices were soaring. The NHS was desperate to cut costs and control the leviathan that healthcare had become.

In 1975, as prenatal screening was on the increase, the benefits of a national screening programme were made abundantly clear:

The advantages gained ..., due to early detection of severe neural-tube defects and other abnormalities, [would lead to] ... a major reduction in the number of cases of spina bifida requiring long-term institutional care. In crude economic terms, the value of the savings in healthcare alone would probably far outweigh any costs of a screening programme.<sup>1</sup>

Coupled with the wider economic concerns of the NHS, Leighton's cost-cutting prophecy was widely believed. Not only were costs to the NHS being estimated—a term that cannot be over-exaggerated in this context—but researchers also alluded to cost savings to society in domains such as education and infrastructure support for future citizens living with disability.<sup>38</sup> However, it was the assumption, made by Leighton and others, that disability was a cost to the nation that leaps out. Here the phrase "would probably far outweigh" suggests that the real cost of disability was yet to be determined, as was also the case in the 1977 collaborative study that at once assured that savings could be made but revealed that this was based on "untested assumptions".<sup>13</sup>

It is clear that the pre-existing understanding of disability was influential in both defending and promoting prenatal screening and termination based on the notion that these were costly and unproductive citizens. The work of Glass and Cove<sup>39</sup> for the DHSS finally confirmed what others had surmised in 1978: that widespread screening could save the public coffers. They suggested that a screening programme would pay for itself in 1 year if it was 95% accurate. Some were less convinced of the amount of savings that could be made, but the desire to save money was too powerful to resist.<sup>40</sup> This desire fed into Meade's vision for the NHS, where preventative action would limit the numbers of sick and save money.<sup>37</sup> Of course, this argument had its flaws as discussed by Cochrane in 1971 and McKeown in 1976, but this idea was both seductive and prevalent.<sup>5</sup>

There was some effort to sound as if financial considerations were not paramount, but it meant little. The above-discussed DHSS document had suggested that although humanitarian arguments to prevent spina bifida were vital, economic costs were worthy of consideration. This was a hollow statement as only the issue of costs was fully addressed. This was in evidence throughout most of the letters and studies already discussed.

The second issue in relation to benefit was the utilitarian notion of happiness. Covered most comprehensively by Chamberlain in 1978, she considered the non-financial side of

the benefit argument. She suggested that the key benefit of screening would be the termination of affected fetuses, delicately described as “averting the birth”. In one sense, benefit was defined as preventing the births of those “who would have survived to live a handicapped life”.<sup>33</sup> In her utilitarian calculus, benefit for society increased in direct proportion to the number of terminations, and the prevention of the perceived suffering of abnormal children. This fell in line with the efficacy debates as Chamberlain understood costs as the potential termination of a healthy child. Thus, benefit is understood in terms of the number of true positives terminated, and cost is understood in terms of the number of false positives terminated. According to Chamberlain, we need simply to weigh these benefits and costs to perform the calculus.

Although she was uncertain as to the efficacy of widespread screening, she had no doubt as to the benefits of preventing spina bifida births:

As with many other screening programmes, it is disappointing, when benefits are estimated on a population basis to find that so many affected pregnancies are likely to escape early detection and termination ... but a reduction of 200 births a year is certainly a worthy objective.<sup>33</sup>

For Chamberlain, the calculus is clear, and benefit clearly outweighed any cost. She even minimises the pain of the cost of terminating a normal fetus when she says:

It is generally assumed that termination half-way through an affected pregnancy causes less upset than a still or neonatal death, and the distress which a severely handicapped child imposes on a family is well documented.<sup>33</sup>

At the heart of such ideas, as Polini<sup>41</sup> suggested in 1978, was the ability to control pregnancy.<sup>41</sup> By harnessing technology, the “problem” of spina bifida could be lessened to create benefit to the individual and society by “diminish[ing] the burden of unhappiness”.<sup>41</sup> Although the *Lancet* collection did not spend much time discussing this position, implicitly, the basis of prevention in relation to benefit was also measured in terms of happiness to the nation.

Cost then was seen by some as a negative term that highlighted the unhappiness brought about by the life of a disabled person, seen especially in terms of long-term financial burden. The one unambiguous moment of dissension came from Ellison-Nash who clearly saw the implications of such arguments. In a strident letter to the editor, he suggested that crude cost analysis did not take into account delineations of impairment. Nor did it reflect upon the “scores of happy useful citizens earning their living who were born with an open spinal defect”.<sup>18</sup> The balancing of these costs and benefits was only possible if a more efficacious screening test could be achieved, with statistical lines drawn at appropriate levels. For most authors, cost was based on evaluating the loss of those “normals” that were terminated, and those “abnormals” who were not terminated; benefit was constructed as the prevention of those who were diseased, or more accurately, as the eradication of the “abnormal” that was embedded in negative connotations of disablement.

## CONCLUSIONS

In this study, we have looked historically at the justification of prenatal screening for spina bifida and although this analysis focuses on spina bifida, it is clear that our conclusions could pertain to other fetal abnormalities. Indeed, the work of Nicolas Wald traversed spina bifida and Down’s syndrome.<sup>30–42</sup> It was

no surprise to see that ethics was labelled in relation to the doctor–patient relationship. What was surprising was conversational tone taken by the authors when discussing the ethics. While ethics was named as the doctor–patient relationship, we have shown that real ethical issues—those discussions around the justification of screening—lay silent, hidden beneath the language of statistical effectiveness and assumed notions of benefit. We have claimed that by looking at extensive sections on efficacy and statistics—by looking at what was contrasted with the ethics and the humanitarian—we see an elaborate justification offered for effective widespread screening programmes geared towards the eradication of spina bifida.

In reality, by stressing the efficacy and benefit, the notion of prevention was justified in quantifiable terms. The benefits of prenatal screening for spina bifida were to save the national coffers, which would increase happiness. Moreover, by reducing the costs associated with disability, and by supporting technology, the ailing economy would also reap benefits.

As Skrabanek has said:

It does not matter what you screen for ... is prevention better than cure? To ask about the ethics of screening, generally aimed to make healthy people healthier, sounds, if not perverse, then definitely suspicious. The fact that screening is a swinging, lucrative business is an incidental phenomenon—a rare example of goodness being rewarded on this earth.<sup>8</sup>

What we have then is a moral calculus, or perhaps better, a complex moral economy, where both monetary and non-monetary benefits were assumed, and where social and scientific progress were linked together in an attempt to create an efficient and effective programme of delivering what we were assured society needed.

The connection between the category of disability and economics was nothing new, but when medicine justifies its actions in terms of the effectiveness of AFP screening towards assumed benefits of prevention of spina bifida, or better, towards the eradication of the costly abnormal, it is clear that we are dealing with a broader notion of justification of action. That which was named the ethical was only part of a much more complex moral economy.

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